Decreased chest mobility in children with spastic cerebral palsy

Murat Ersöz, Barın Selçuk, Ramazan Gündüz, Aydan Kurtaran, Müfit Akyüz

1st Physical Medicine and Rehabilitation Clinic, Ankara Physical Medicine and Rehabilitation Training and Research Hospital of Ministry of Health, Ankara, Turkey

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Consequences of neuromuscular impairment may lead to lung damage and reduced lung function in children with cerebral palsy (CP). The purpose of this study was to evaluate chest mobility by means of chest expansion (CE) measurements in patients with spastic CP. Chest circumference at maximal voluntary inspiration (Cinsp) and at maximal voluntary expiration (Cexpir) and CE (the difference between Cinsp and Cexpir) were measured in 56 consecutive inpatients with spastic CP and in 40 healthy children. CE was significantly decreased ($p < 0.001$) and Cexpir was increased ($p < 0.02$) in the CP group, while mean Cinsp values were not statistically different ($p > 0.05$). The difference between CP patients and controls with respect to CE was becoming more prominent in older children.

As chest mobility is decreased in spastic CP patients, early initiation of pulmonary rehabilitation, which may improve and maintain chest mobility and respiratory function, seems reasonable in this patient group.

Key words: cerebral palsy, chest expansion, chest circumference, respiratory dysfunction.

Cerebral palsy (CP) is a collection of diverse syndromes characterized by disorders of movement and posture caused by nonprogressive injury to the immature brain[1]. While CP itself does not directly cause airway or parenchymal lung dysfunction, consequences of neuromuscular impairment may lead to lung damage and reduced lung function[2-6]. Poor nutritional status, drooling, aspiration, gastroesophageal reflux, impairment of airway clearance by muscular weakness or incoordination and poor pulmonary reserve (due to chest wall or spine deformity and spasticity) increase the risk of significant morbidity and mortality from respiratory infections[5-9]. Those individuals who were premature infants or who had prolonged neonatal courses may also have residual chronic lung disease contributing to their pulmonary problems[5,10]. Although survival rates of mild and moderately disabled CP patients are not much lower than those of unaffected children and about half of severely disabled children survive to age of 20, lung disease remains an important cause of morbidity and mortality for this patient group[11-13].

The aim of this study was to evaluate chest mobility by means of chest expansion measurements in spastic CP patients and in subgroups according to type of involvement, ambulation level, mental status and grade of spasticity, which would help to gain a better understanding of the role of respiratory muscle weakness, incoordination, spasticity, thoracic cage limitations, mental status and physical activity level on respiratory dysfunction. Marked decreases in the vital capacity for subjects with dyskinetic and spastic CP and positive effects of various exercise programs on vital capacity have been reported previously[2-4]. We believe that the findings of our study may be helpful in rehabilitative efforts focused on respiratory dysfunction, especially in choosing the appropriate exercises.

Material and Methods

Fifty-six consecutive children with spastic CP with a mean age of 6.7±2.0 years (range: 3-12) and 40 healthy control subjects with a mean age of 6.8±1.8 years (range: 4-11) were evaluated in the study. All children with CP

were inpatients hospitalized for rehabilitation. None of the children had any apparent evidence of intrinsic lung disease. Body weight, height and tibial length of the right leg, which have been reported as a reliable and valid proxy for stature in children with cerebral palsy up to 12 years of age[14], were recorded for each subject. Type of involvement (quadriplegic, triplegic, diplegic or hemiplegic)[1], mental status (normal or mental retardation), grade of spasticity according to Ashworth scale (slight, mild, moderate, severe), level of ambulation (non-ambulatory, ambulatory with orthosis and/or assistive device, ambulatory without orthosis and assistive device) and presence of scoliosis and kyphosis were noted for CP patients.

Chest circumference at maximal voluntary inspiration (Cinsp) and at maximal voluntary expiration (Cexpir) and chest expansion (CE) (the difference between Cinsp and Cexpir) were measured in sitting position using a tape measure marked in 0.1 cm increments at the level of the fourth intercostal space by the first author. All children were informed about the examination and were asked to exhale as much as possible and hold the position for Cexpir measurements and to take as deep a breath as possible and to hold it for Cinsp measurements[15]. Only the subjects who could cooperate throughout the examination were included in the study. The highest value of Cinsp and the lowest value of Cexpir in three attempts were recorded for all children and the difference between Cinsp and Cexpir was recorded as CE.

SPSS software version 8.0 was used for data analysis. The data were expressed as the mean and standard deviation or as frequencies when appropriate. The spastic CP and control groups were compared by independent samples t-test for age, body weight, height, tibial length, Cinsp, Cexpir, CE and by chi-square test for gender. One-way ANOVA and Tukey Honestly Significant Difference (HSD) analysis were conducted to document the differences among CP subgroups according to type of involvement, mental status, ambulation level and grade of spasticity and the control group. Associations between grade of spasticity and Cinsp, Cexpir, CE were investigated by Spearman correlation test. A level of $p < 0.05$ was used as a cut-off level for statistical significance.

Results

The distribution of spastic CP patients according to type of involvement, mental status, ambulation level and grade of spasticity and the frequencies of scoliosis and kyphosis in CP patients are presented in Table I. None of the patients had

Table I. Distribution of CP Patients According to Type of Involvement, Mental Status, Ambulation Level, Grade of Spasticity and Presence of Scoliosis and Kyphosis

	n (56)	%
Type of involvement		
Quadriplegic	20	35.7
Triplegic	12	21.4
Diplegic	21	37.5
Hemiplegic	3	5.4
Mental status		
Normal	43	76.8
Mental retardation	13	23.2
Grade of spasticity (Ashworth)		
Slight (1)	8	14.3
Mild (2)	38	67.9
Moderate (3)	10	17.9
Severe (4)	0	0
Ambulation level		
Non-ambulatory	12	21.4
Ambulatory with orthosis and/or assistive device	42	75
Ambulatory without orthosis and assistive device	2	3.6
Scoliosis		
Absent	46	82.1
Postural	7	12.5
Present	3	5.4
Kyphosis		
Absent	47	83.9
Postural	7	12.5
Present	2	3.6

apparent chest deformity. Results of comparison of spastic CP patients and control subjects for demographic and clinical parameters are presented in Table II. Although the groups were similar with respect to age and gender, there were statistically significant differences between groups with respect to height, weight, tibial length, Cexpir and CE. Height, weight, and tibial length were significantly decreased in children with CP. CE was significantly decreased and Cexpir values were increased in CP patients (Table II and Fig. 1). The changes in CE by age in spastic CP and control groups are shown in Figure 2. The difference between CP patients and controls with respect to CE was becoming more prominent in older children, as can be seen in Figure 2.

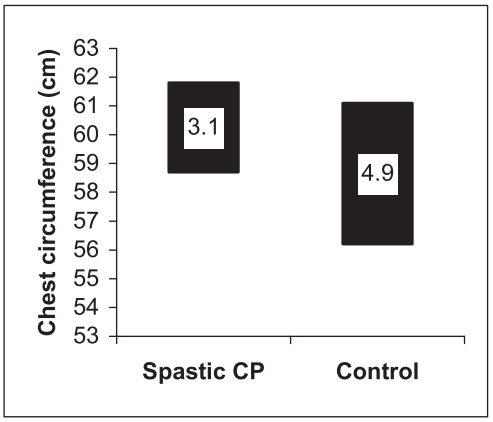

Fig. 1. Chest expansion (CE) in spastic CP and control groups (Upper borders of the bars represent Cinsp and lower borders represent Cexpir, and the values given in the bars represent CE).

Table II. Comparison of Results of Spastic CP and Control Groups for Gender, Age, Body Weight, Height, Tibial Length, Cinsp, Cexpir, and CE [Values for CP Subgroups are Given in the Lower Part of The Table (mean ± SD)]

	Gender (M/F)	Age (years)	Height (cm)	Weight (kg)	Tibial length (cm)	Cexpir (cm)	Cinsp (cm)	CE (cm)
CONTROLS (n:40)	28/12	6.8±1.8	116.4±11.3	21.0±5.2	24.3±3.4	56.2±4.7	61.1±5.4	4.9±1.2
CEREBRAL PALSY (n:56)	43/13	6.7±2.0	106.6±15.9	17.6±4.8	22.7±3.0	58.7±5.2	61.8±5.3	3.1±0.9
P	0.455	0.819	0.001	0.001	0.012	0.018	0.519	<0.001
CP SUBGROUPS								
Type of involvement								
Quadriplegic (n:20)	15/5	7.0±2.0	102.6±23.2	16.8±5.1	22.3±3.3	57.9±5.0	60.7±5.0	2.9±0.9
Triplegic (n:12)	11/1	6.9±1.7	112.2±9.9	19.7±5.8	23.3±3.1	60.0±6.7	63.4±7.1	3.4±0.6
Diplegic (n:21)	15/6	6.3±2.1	106.5±9.3	17.2±4.0	22.6±2.7	58.8±4.8	62.1±4.7	3.2±1.1
Mental status								
Normal (n:43)	31/12	6.2±2.2	107.0±17.0	18.1±5.3	22.8±3.3	59.0±5.5	62.3±5.6	3.2±0.9
Mental retardation (n:13)	12/1	6.8±0.8	105.5±5.9	16.0±2.4	22.1±1.7	57.6±4.0	60.4±4.0	2.8±0.8
Ambulation level								
Non-ambulatory (n:12)	10/2	5.8±1.5	101.0±7.2	14.3±2.1	20.5±1.8	55.9±3.1	58.6±3.5	2.8±0.7
Ambulatory with orthosis and/or assistive device (n:42)	32/10	6.9±2.1	107.8±17.6	18.4±5.1	23.2±3.0	59.5±5.5	62.7±5.5	3.2±0.9
Grade of spasticity (Ashworth)								
Slight (1) (n:8)	6/2	6.1±0.8	108.5±5.1	17.5±2.4	22.8±1.6	57.2±4.7	61.0±4.3	3.8±1.2
Mild (2) (n:38)	29/9	6.7±2.3	105.9±19.0	17.7±5.6	22.7±3.3	59.1±5.6	62.1±5.9	3.0±0.7
Moderate (3) (n:10)	8/2	6.9±0.9	108.0±6.3	17.2±2.9	22.5±2.6	58.5±4.2	61.3±4.1	2.8±1.0

CP: Cerebral palsy. Cexpir: Maximal voluntary expiration. Cinsp: Maximal voluntary inspiration. CE: Chest expansion.

The results of subgroups according to type of involvement, mental status, ambulation level and grade of spasticity are presented in Table II. There were statistically significant differences in one-way ANOVA among subgroups according to type of involvement and controls with respect to height ($p<0.005$), weight ($p<0.01$) and CE ($p<0.001$). Hemiplegic patients were not included in this analysis because of the limited patient number. Post hoc analysis revealed that quadriplegic children had lower height ($p<0.005$) and weight ($p<0.02$) values than controls, diplegic patients had lower weight values than controls ($p<0.04$) and quadriplegic, triplegic, and diplegic CP patients had decreased CE when compared with controls ($p<0.001$). There were no significant differences among quadriplegic, triplegic and diplegic CP subgroups in any parameter ($p>0.05$).

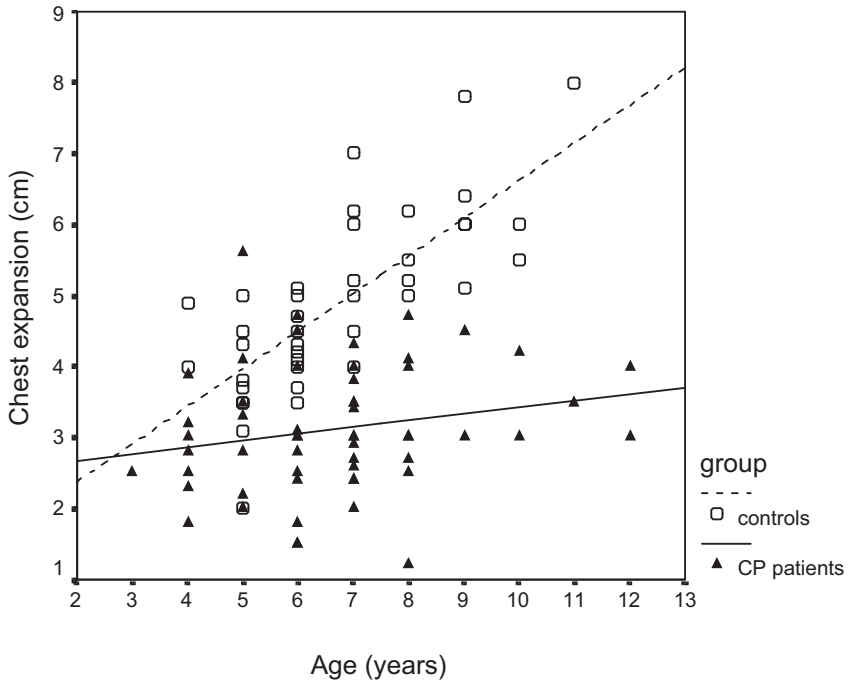

Fig. 2. The changes in chest expansion by age in spastic CP and control groups.

According to mental status, there were statistically significant differences among controls and subgroups with respect to height ($p<0.006$), weight ($p<0.004$), CE ($p<0.001$) and Cexpir ($p<0.05$). Post hoc analysis revealed that children with mental retardation had lower height ($p<0.05$) and weight ($p<0.008$) values than controls; CP patients with normal mental status had lower height ($p<0.01$), weight ($p<0.03$) and higher Cexpir ($p<0.04$) values than controls; and both mentally retarded and CP patients with normal mental status had decreased CE ($p<0.001$). There were no significant differences between mentally retarded and mentally normal CP subgroups in any parameter ($p>0.05$).

There were statistically significant differences among controls and subgroups according to ambulation level with respect to height ($p<0.003$), weight ($p<0.001$), tibial length ($p<0.002$), Cexpir ($p<0.007$) and CE ($p<0.001$). Patients who were ambulatory without orthosis and/or assistive device were not included in this analysis because of the limited patient number. In post hoc analysis non-ambulatory children had lower height ($p<0.005$), weight ($p<0.001$) and tibial length ($p<0.002$) values than controls; CP patients who were ambulatory with orthosis and/or assistive device had lower height ($p<0.02$) and higher Cexpir ($p<0.02$) values than controls; and CE values were decreased in both CP groups ($p<0.001$). Non-ambulatory CP patients had significantly lower weight ($p<0.04$) and tibial length ($p<0.03$) values than ambulatory patients with orthosis and/or assistive device. There were no significant differences between subgroups with respect to CE, Cexpir and Cinsp ($p>0.05$).

Statistically significant differences were observed among subgroups according to grade of spasticity and controls with respect to height ($p<0.02$), weight ($p<0.02$) and CE ($p<0.001$). There were no patients with severe spasticity. Post hoc analysis revealed that children with mild spasticity had lower height ($p<0.01$) and weight ($p<0.03$) values than controls. Patients with mild and moderate spasticity ($p<0.001$) and patients with slight spasticity ($p<0.05$) had decreased CE. There were no significant differences among subgroups with respect to any parameter ($p>0.05$).

In Spearman correlation test, a weak negative correlation was observed between grade of spasticity and CE ($r= -0.283$, $p<0.05$) in spastic CP patients, but there were no correlations between grade of spasticity and Cinsp or between grade of spasticity and Cexpir.

Discussion

The results of this study showed that chest mobility as evaluated by CE was decreased in spastic CP patients when compared with normal controls of similar age and gender. CE was significantly decreased in all CP subgroups according to type of involvement, mental status, grade of spasticity and level of ambulation. Although lower CE values were observed in quadriplegia, mental retardation, moderate spasticity (there were no patients with severe spasticity) and non-ambulatory subgroups, the differences among subgroups did not reach statistical significance. For chest expansion, normal functions of nervous system, respiratory muscles and costovertebral joints are needed[15,16]. As CP does not have articular involvement, the limited chest mobility may be due to impaired neuromotor control and incoordination, weakness, spasticity and secondary changes in the respiratory muscles[2-4,16-18]. The inability of the respiratory muscles to adequately increase and decrease the volume of the thoracic cavity may result in stiffening of the costovertebral joints, which may also decrease chest expansion[16]. The abnormal breathing patterns seen in CP patients because of the problems mentioned above are shallow breathing (rapid series of low amplitude excursions coupled with low vital capacity) and reversed breathing (the thorax makes inspiratory excursions while the abdomen makes expiratory excursions, or vice versa)[3,16,18]. Breathing by these abnormal patterns for a long period may further limit chest mobility by shortening of respiratory muscles and stiffening of costovertebral joints.

In the review of the literature, we could not find any study that investigated chest mobility in children with CP, but marked decreases in vital capacity have been reported in this patient group[2-4]. In these studies, mean vital capacities were 23-67% of the predicted normal values[2-4]. Vital capacity is defined as maximal total volume excursion that includes inspiratory capacity and expired reserve volume[19]. As chest expansion is the difference between maximal inspiration and maximal expiration, it can be speculated that the limited chest expansion observed in our study may have a major role in decreased vital capacity seen in CP patients, but detailed studies are needed to show this possible relationship.

Additional interesting findings of our study were slightly increased mean Cinsp and significantly increased mean Cexpir values observed in the CP group when compared with controls. As CP patients in our study had significantly decreased height, weight and tibial length, reflecting undernutrition and growth failure which are common in children with CP[7], the observed slightly increased Cinsp and significantly increased Cexpir instead of decreased Cinsp and Cexpir were surprising. These findings revealed a tendency to development of barrel chest in spastic CP patients, which is a chest deformity seen in chronic obstructive lung diseases and is a sign of chronic air-trapping[15,20]. One-second forced expiratory volume test (FEV1) results, which are used for evaluation of expiratory function, were found to be low in CP patients in one study[2], whereas they were normal in another[3]. It has been reported that an approximately 50% increase in FEV1% vital capacity values could be obtained with an adaptive seating system which corrected the posture of the thorax and head[21]. Marked increases in expiratory time and vital capacity were also observed by this adaptive seating system and these changes were attributed to the unobstructed airway obtained by this system[21]. According to some authors, children with CP behave like patients with obstructive lung disease although they have no signs of obstruction[2,21]. This may be the result of neuromotor disturbance which caused inability to force the thorax back to normal size[2] and/or may be due at least in part to some degree of obstruction of the upper airways because of the incorrect posture of the thorax, neck and head[21]. Incoordinated laryngeal opening with diaphragmatic activity may be another possible cause of airway obstruction[18]. The significantly increased mean Cexpir and the observed higher mean Cinsp than expected in our study can be explained by inability to force the thorax back to normal size and by chronic hyperinflation, which may be the result of incomplete expiration. We believe that these findings supported the possibility of obstructive lung impairment in spastic CP patients. Furthermore, there were postural abnormalities in 16/56 (28.6%) of CP patients in our study, which could possibly cause upper airway obstruction as explained[21]. Finally, aspiration with or without gastroesophageal

reflux may be another causative factor for obstructive respiratory dysfunction in CP patients, but new studies are needed for clarification of the presence of obstructive respiratory dysfunction in children with spastic CP and for determination of the underlying mechanisms.

Another important finding to mention is significantly decreased CE in all spastic CP patients independent of the type of involvement. Although marked respiratory muscle involvement was not expected in diplegic patients, CE was also decreased in this subgroup. The possible explanation of this uniform decrease in CE observed in all CP subgroups may be the limited physical activity. In our study, 13 of 56 patients were non-ambulatory, 42 were ambulatory with orthosis and/or assistive device, 2 were ambulatory without orthosis and/or assistive device and all patients had very limited physical activity. None of the patients was able to run or participate in sports as normal children do. This finding suggested that increased respiratory demand as a result of intensive physical activity has a major role in the development of normal chest mobility, but this possible relation should be investigated by further studies. Increased physical work capacity in children with CP has been reported after physical training programs without any negative side effects[22]. Effect of such training programs on chest mobility may be another future field of investigation.

As lung disease is an important cause of morbidity and mortality for CP patients[13], evaluation and management of respiratory dysfunction should be recognized as part of the routine rehabilitation. Respiratory muscle strengthening exercises, especially for muscles of expiration, which would help to decrease the thorax to normal size as well as facilitate construction of effective cough, should be included in the rehabilitation program. Chest expansion exercises, which include maximum inspiration and expiration, will prevent stiffening of costovertebral joints and shortening of respiratory musculature. Strengthening of trunk and neck muscles coupled with posture exercises and adaptive seating systems may improve posture, diminish the obstruction of the upper airway because of bad posture and improve respiratory function. Breathing exercises will be helpful in achieving effective inspiration and expiration, in improving coordination and in elimination of abnormal breathing patterns seen in children with CP. Aquatic exercises, when available, may be beneficial for CP patients because of the hydrostatic pressure of the water, which provides increased pressure on the chest - they may help effective expiration and to decrease thorax volume to normal size. Finally, regular exercises for intensive physical activity that may improve chest mobility can be conducted. Aquatic exercises[4], bicycle[22], arm ergometry or programs of quicker and slower motions[22] can be chosen for this purpose according to the patient's ability.

In conclusion, chest mobility as evaluated by CE is decreased in spastic CP patients, which may interfere with respiratory function. The difference between CP patients and controls becomes more prominent in older children and there is a tendency to development of barrel chest. These findings raise the need of early initiation of pulmonary rehabilitation, which may improve and maintain chest mobility and respiratory function, prevent development of chest deformity and, as a consequence, decrease the morbidity and mortality due to lung disease.

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